Castleman's Disease of the Chest Wall

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Abstract: Primary Castleman's disease of the chest wall is unusual. Furthermore, such tumors arising from a surgical wound are extremely rare. We report a 33-year-old female with a history of a thoracic surgery at 5 years of age. A round, homogenous 4 × 3.5-cm mass protruded into the thoracic cavity on the posterior portion of the previous posterolateral incision. The tumor was completely removed, with combined rib resection. The resected specimen showed Angiofollicular Lymph Node Hyperplasia (Castleman's disease), hyaline-vascular type. No recurrence has been found for 10 years. This is the first report of primary chest wall Castleman's disease arising from the surgical wound.

Key Words: Castleman's disease, Chest wall, Trauma.

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Primary Castleman's disease (CD) of the chest wall is unusual.[1] We report an extremely rare case of Angiofollicular Lymph Node Hyperplasia (CD) arising from a surgical wound of the chest wall. In addition, we discuss the etiology and treatment of CD.

CASE REPORT

A 33-year-old female was referred to our hospital for a 2-month history of left shoulder pain. She had undergone closure of a patent ductus arteriosus through posterolateral incision at the age of 5 years. Chest radiograph showed a round, homogenous 4 × 3.5-cm mass with sharp margin in the left upper lung field (Figure 1). Chest computed tomography showed homogenous soft-tissue-attenuating mass with sharp margin (Figure 2). We performed a thoracotomy through the fifth intercostal space, following left posterolateral incision. Dense adhesion was found between the lung and posterior parietal pleura at the level of the third and fourth ribs, which corresponded to the previous surgical wound. A hen's egg-sized elastic hard mass was palpable through the corresponding lung. Adhesion between the tumor and the visceral pleura of the lung was released, and the tumor was completely removed with combined resection of the third and fourth ribs. The patient experienced no postoperative complication and has been followed up for 10 years without relapse.

On gross examination, the tumor was found to be an encapsulated brown mass measuring 3.5 × 4.0 × 3.0 cm, stuck into the intercostal space. Sectioning of the mass revealed a yellow solid surface, without invasion to surrounding tissue (Figure 3). Microscopic features showed the presence of numerous germinal centers interspersed in a population of mononuclear cells and numerous capillaries without sinusoid (Figure 4). At the germinal center, a small vessel with a thickened, hyalinized wall extended into the midportion. These appearances were consistent with Angiofollicular Lymph Node Hyperplasia (CD), hyaline-vascular type, originating from the chest wall.

Comment

CD, Angiofollicular Lymph Node Hyperplasia, is one of the atypical lymphoproliferative diseases. CD located in the chest wall is rare; only, 11 cases have been reported in the English and French literature.[1] Furthermore, there have been no cases arising from surgical wounds. We report here an exceedingly rare case of CD arising from a surgical wound of the chest wall.

CD is classified into unicentric and multicentric on the basis of clinical appearance[2] and into hyaline-vascular type and plasma cell type on the basis of microscopic manifestation.[3] Most CD located in the chest wall are unicentric and hyaline-vascular type.[1] Like most cases, ours was unicentric and hyaline-vascular type.

The etiology of CD is unknown, although several theories have been presented. Chronic inflammation, hamartomatous process, immunodeficiency state, and autoimmunity have been proposed as etiologies of this disease.[4] In multicentric CD, Kaposi sarcoma-associated herpesvirus is related to its tumorigenesis.[5] We here demonstrate a case of CD arising from a surgical wound. Gilliam and Woods[6] pointed out that lymphoid hyperplasia, including CD, might represent exaggerated reactions to diverse external antigens including trauma in the skin. In our patient, the wound stimuli such as remnant thread or infection around the wound, might have been a pathogenesis of CD.

Surgery is essential for unicentric type CD. Bowne et al.[4] reported that complete surgical resection in 10 unicentric CD patients achieved no recurrence at least for 9 to 37 months. We successfully treated our patient with complete resection, and have observed no complications or relapses until now. However, those authors pointed out that partial resection, radiotherapy or observation alone might be an alternative to complete resection, since three patients with

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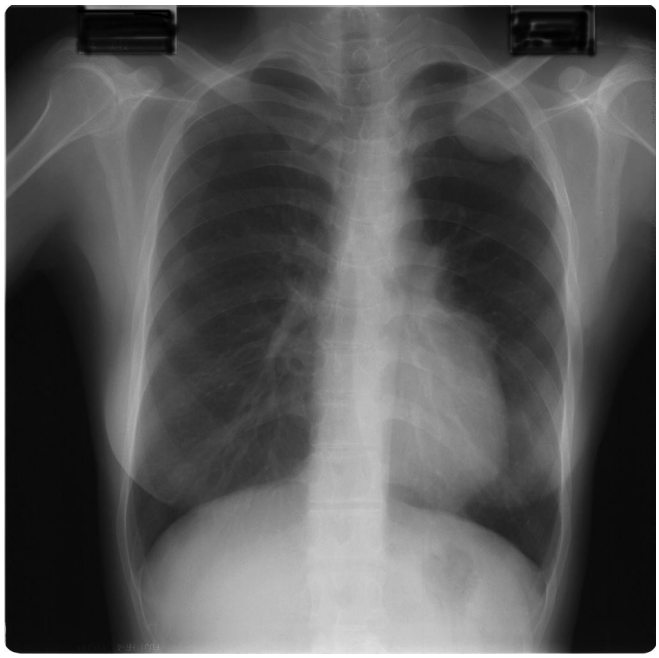

FIGURE 1. Posteroanterior chest radiograph demonstrating homogenous tumor close to deformed third rib of left chest wall.

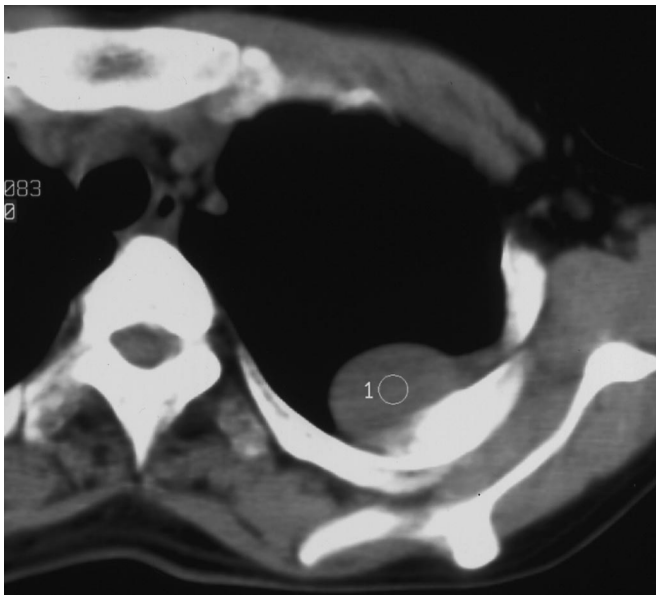

FIGURE 2. Chest computed tomography demonstrating homogenous soft-tissue-attenuating mass with sharp margin.

remnant lesion remained asymptomatic for at least 12 months. Neuhof and Debus[7] reported that 4 of 5 unicentric CD patients were successfully treated with radiation. Radiation may be another treatment option. Multicentric CD is defined as a systemic disease. Many such patients have a poor prognosis, though multimodality therapy including radiotherapy and chemotherapy have been administered.[5] Recently, a unicentric, non-HIV-associated hyaline–vascular type CD patient was successfully treated with anti-CD20 therapy.[8] This may be another alternative therapy for tumor not completely resected.

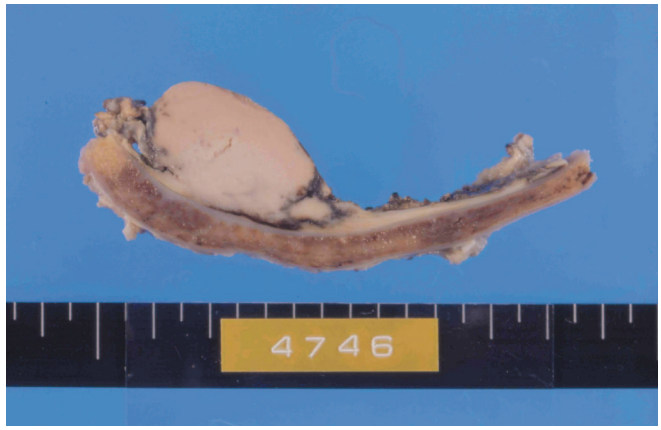

FIGURE 3. Macroscopic appearance of oval solid tumor with chest wall. Tumor was encapsulated and did not invade surrounding tissues.

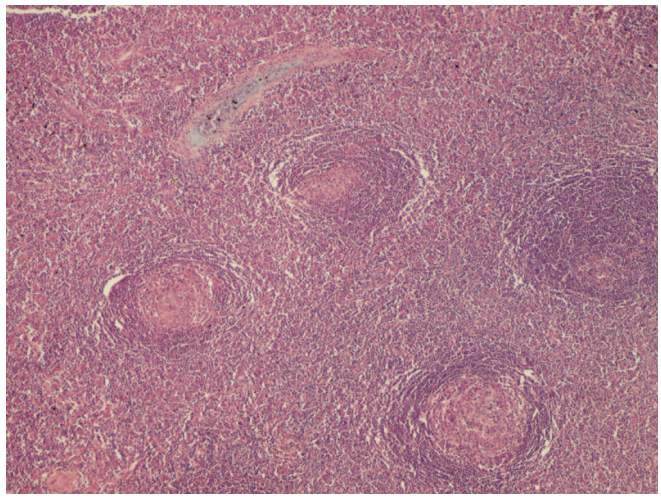

FIGURE 4. Microscopic appearance of resected tumor (Haematoxylin and Eosin [HE], ×80). Hyperplastic lymphoid follicle has small hyaline vascular structure and is surrounded by cuff of small lymphocytes in concentric "onion skin" layers.

We demonstrate a case of CD arising from a surgical wound of the chest wall, and suggest that its etiology might be the surgical wound. We believe that this is the first report in the literature of CD arising from the surgical wound.

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